CASE REPORT

Scalp metastasis from atypical meningioma: A case report and literature review

Saad Javed¹ Amina Khan² | Ayesha Khalid² | Gianluca Scalia³ Giuseppe Emmanuele Umana⁴ | Ashraf Mahmood² | Eesha Yaqoob⁵ | Bhavya Pahwa⁶ | Bipin Chaurasia⁷

¹Department of Neurosurgery, Rawalpindi Medical University, Rawalpindi, Pakistan

²Department of Neurosurgery, Rawalpindi Medical University Rawalpindi, Rawalpindi, Pakistan

³Neurosurgery Unit, Department of Head and Neck Surgery, Garibaldi Hospital, Catania, Italy

⁴Department of Neurosurgery, Gamma knife and Trauma Center, Cannizzaro Hospital, Catania, Italy

⁵Department of Public Health, Health Services Academy, Ministry of National Health Services Regulations and Coordination, Government of Pakistan, Islamabad, Pakistan

⁶University College of Medical Sciences and GTB Hospital, Delhi, India

⁷Department of Neurosurgery, Neurosurgery Clinic, Birgunj, Nepal

Correspondence

Bipin Chaurasia, Department of Neurosurgery, Neurosurgery Clinic, Birgunj, Nepal. Email: trozexa@gmail.com

Key Clinical Message

Scalp metastasis from atypical meningioma, though rare, underscores the importance of meticulous surgical techniques to prevent tumor cell implantation. Early detection and comprehensive management, including surgery and adjuvant therapy, are crucial for optimal outcomes.

Abstract

Meningiomas are tumors of the meninges that originate in the arachnoid layer from arachnoid cap cells. Atypical meningiomas, classified as WHO grade 2 tumors, tend to metastasize and recur if not surgically managed properly. Scalp metastasis is a rare occurrence that presents as a subcutaneous elevation. A 33-year-old patient presented with a complaint of a constant, dull pressure headache persisting for the past 12 months, exacerbated by exertion, along with seizures and neuropsychiatric symptoms. The patient had no significant medical history but had undergone surgery 4 years ago for a WHO grade 2 meningioma. The current brain MRI revealed a dural tail sign, along with masses on both the left and right sides of the frontal lobe, extending to involve the skin on the forehead and scalp. The patient underwent surgical resection and adjuvant radiation therapy. At the 12-month follow-up, no neurological deterioration or tumor recurrence was observed. A literature review on scalp metastasis in patients with atypical meningioma was also conducted, including eight articles published up to September 2023. The mechanism of metastasis development appears to be consistent in all eight reported cases, involving the implantation of tumor cells during resection. Therefore, there is a critical need for meticulous intra- and postoperative surgical techniques to prevent such implantation.

KEYWORDS

atypical, meningioma, metastatic recurrence, radiation therapy, scalp metastasis, surgical resection

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made. © 2024 The Authors. Clinical Case Reports published by John Wiley & Sons Ltd.

1 | INTRODUCTION

Meningiomas account for a substantial proportion of benign tumors (53.3%) and primary brain tumors (38.3%) in the United States.¹ According to the World Health Organization (WHO) classification, meningiomas are categorized into three grades: benign (grade 1, 89.30%), atypical (grade 2, 5.90%), and anaplastic (grade 3, 4.80%) meningiomas.¹ Atypical meningiomas, classified as WHO grade 2, are characterized by their metastatic potential and proclivity for recurrence, presenting significant challenges in their surgical management and treatment.²

Meningioma metastasis primarily occurs through general circulation or iatrogenic distribution.³⁻⁴ Skin metastasis from internal organ cancers accounts for 5%–9% of cases.⁵ Metastasis from malignant meningiomas is exceedingly rare, further underscoring the rarity of scalp metastasis. One established pathology is iatrogenic metastasis resulting from the resection of atypical meningiomas, emphasizing the importance of meticulous surgical techniques to minimize the risk of metastatic recurrence.⁶ This study presents a case of scalp metastasis from atypical meningioma and provides a comprehensive literature review on the topic.

2 | CASE HISTORY/ EXAMINATION

A 33-year-old female patient presented to our tertiary care hospital in 2017 with a complaint of persistent headaches for 12 months. She described the headaches as dull and pressure-like, worsened by coughing, sneezing, and straining. Additionally, she experienced occasional generalized tonic-clonic seizures and exhibited behavioral changes such as depression, memory disturbances, anxiety disorder, and paranoid ideation. The patient had no other significant medical history.

3 | METHODS (DIFFERENTIAL DIAGNOSIS, INVESTIGATIONS AND TREATMENT)

Brain magnetic resonance imaging (MRI) revealed a large meningioma located in the left frontal convexity, and histopathology confirmed it as a WHO grade 2 meningioma. The recommended treatment plan involved radiation therapy and chemotherapy, followed by surgical resection of the meningioma. However, the patient only underwent tumor excision at another tertiary care hospital and did not receive radiation or chemotherapy, resulting in tumor recurrence. Four years later in 2021, the patient returned to our hospital with similar complaints, along with tumor involvement on the left side of the forehead skin and scalp. On physical examination, a fixed, firm, tender lesion measuring approximately 10×12 cm on the left side and 7×7 cm on the right side was observed (Figure 1A). No surrounding lymph nodes were affected, and there were no additional lesions on other parts of the body. CT scan showed bone erosion (Figure 1B). Brain MRI showed a broad-based intra- and extra-axial lesion involving the frontal lobe, with associated mass effect, scalp erosion, and a "dural tail sign" (Figure 1C).

To address the recurrence, the patient underwent tumor excision via a left frontal craniotomy in the elective operating room of the neurosurgery department. During the procedure, the eroded frontal bone was also removed, and frozen sections were examined urgently in the emergency department to ensure a positive margin of tumor resection. After approximately 6h, a "Simpson grade I" resection of the tumor was achieved. To reconstruct the skull, titanium mesh placement was performed in collaboration with the plastic surgery department, followed by skin grafting. Immediate postoperative brain CT scan showed a complete removal and good cosmetic results (Figure 2A). Histopathological analysis revealed a meningioma with zones of increased cellularity, necrosis, and pleomorphism, consistent with the diagnosis of an atypical meningioma (Figure 2B).

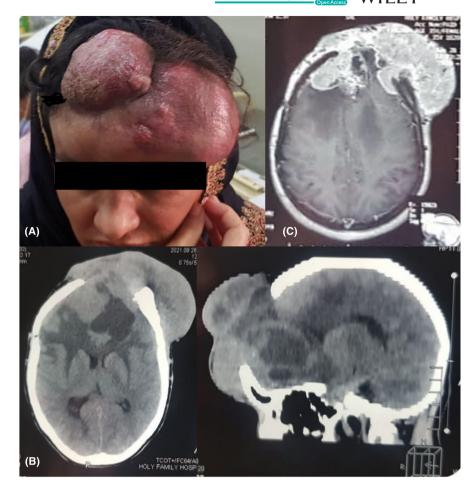
4 | CONCLUSION AND RESULTS (OUTCOME AND FOLLOW-UP)

Postoperatively, radiation therapy was administered to the excision site with wide margins. The patient experienced immediate improvement in her symptoms and remained neurologically stable. Follow-up MRI at 12 months showed no signs of tumor recurrence.

5 | DISCUSSION

A literature review was conducted to compile all case reports on scalp metastasis resulting from atypical (WHO grade 2) meningioma published up to September 2023. The search strategy included keywords such as "WHO grade 2 meningioma," "atypical meningioma," "metastatic meningioma," "malignant meningioma," and "scalp metastasis" or "skin metastasis." Due to the rarity of this condition, all eight studies included in the review were case reports. In all reported cases, patients underwent resection and received radiotherapy, resulting in a 100% survival rate at a mean follow-up of 18 months.

FIGURE 1 Image showing the extensive scalp metastatic lesion due to atypical meningioma (A). Preoperative axial and sagittal brain CT scan images showing bone and skin involvement (B). Preoperative T1-weighted MRI sagittal sequence showing tumor invasion and aggressiveness (C).



Meningiomas account for approximately 15.60% of all primary CNS tumors treated in Pakistan.⁷ The vast majority of meningiomas are nonmalignant (about 90%).⁸ Malignant meningiomas represent roughly 0.4% of all CNS tumors in the United States.⁹ Atypical meningiomas, classified as WHO grade II, exhibit a characteristic tendency for recurrence.¹⁰ The incidence of atypical meningiomas, along with meningiomas in general, is notably higher among female patients both globally¹¹ and in Pakistan.¹²

Scalp skin metastasis from atypical meningioma is an exceptionally rare clinical phenomenon, with only eight reported cases in the literature, making the presented case the ninth and the second from the same country (Table 1). The plausible mechanism underlying metastasis is the dispersion of meningioma cells during surgical resection. Other factors associated with metastatic meningiomas include reoperations, immunosuppression, radiation therapy, and a protracted course of the surgical incision with a cerebrospinal fluid fistula.¹³⁻¹⁶ Knowledge of these factors is valuable for neurosurgeons, despite the rarity of scalp metastasis in meningiomas. The median time interval between diagnosis and scalp metastasis presentation was 44.5 months, primarily explaining the iatrogenic seeding

of cells. The presence of large subcutaneous lesions in our patient suggests the potential presence of cell debris and an invisible tumor at the time of the initial surgery, contributing to implantation metastasis. Surgery has been shown to facilitate the growth of micrometastases at distant sites.¹⁷⁻¹⁸ The resection of the primary tumor may disrupt the inhibitory control exerted by the primary tumor, known as "concomitant tumor resistance," thereby slowing the growth of dormant metastases.¹⁸⁻¹⁹ Additionally, the local extracellular environment, such as the rich vascular supply of scalp wounds, can provide essential nutrients and growth factors to disseminated tumor cells following surgical resection of a primary mass. Surgeons can take precautionary measures to minimize implantation metastasis risk during surgery, such as preserving tumor integrity, conducting piecemeal resection to prevent intraoperative tumor fragment dispersion, isolating equipment that has come into contact with the tumor to avoid cross contamination, and repetitively rinsing exposed tissues with saline.²⁰

The patient underwent surgery consistent with Simpson Grade I, involving excision of the skin patch, bone removal, and affected dura. Literature has consistently shown that complete tumor resection remains

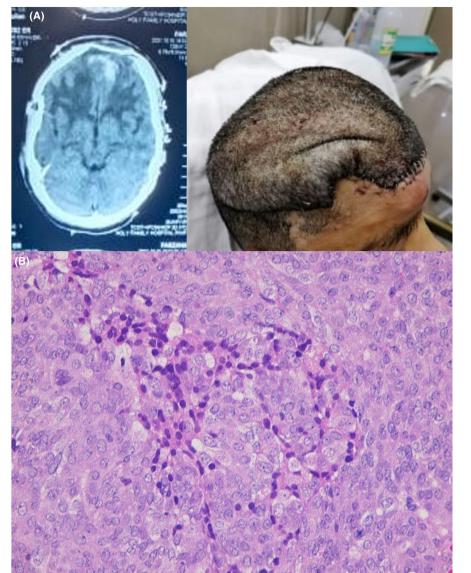


FIGURE 2 Postoperative day-1 brain CT scan after Cranioplasty and postoperative image of patient's head after skin grafting (A). Histopathology – hematoxylin–eosin staining (×100): The tumor cells are arranged in a lobular or nest-like shape, with a vortex-like structure in the local area. The tumor nucleus is of medium size, mitotic images are (>4/10 HPF), and necrosis is seen in the local area (B).

a significant prognostic factor, even when considering other variables like adjuvant radiation therapy and the updated WHO classification of 2016. The extent of resection (Simpson grading) remains a strong prognostic indicator, with Simpson Grade I resections yielding more favorable outcomes.²¹⁻²² Recent evidence suggests the utility of 5-aminolevulinic acid (5-ALA) fluorescenceguided bone resection in infiltrating meningiomas, aiding in the identification of tumor remnants and reducing meningioma recurrence rates.²³ Cranioplasty with a titanium mesh offers a stable and cosmetically pleasing solution for extensive bone defects, as seen in this patient with extensive osseous involvement following tumor surgery.

The literature review suggested a median interval of 41.50 months, with shorter intervals for higher-grade (WHO grade II and III) meningiomas, indicating earlier

presentation of metastasis in high-grade cases.²⁰ This underscores the importance of close follow-up for postoperative patients with WHO grade II and III meningiomas. Due to the scarcity of metastatic meningiomas, there are currently no established treatment protocols. The patient in this case underwent chemoradiotherapy postoperatively, consistent with the treatment approach in most of the reviewed cases. Gamma knife radiosurgery (GKRS) has been used successfully for the treatment of residual scalp metastasis, achieving complete recovery without neurological deficits or recurrence.²⁴ GKRS has also demonstrated efficacy as a primary treatment modality for atypical meningiomas.²⁵ Postoperative radiotherapy has been associated with improved local control, progression-free survival, and is considered an effective treatment for postoperative metastatic meningioma patients.²⁶⁻²⁷

Author, year	Age/ Gender	Time interval Number of (months) surgeries in	Number of surgeries in total	Surgery for the metastasis (Y/N)	Adjuvant radiotherapy	Follow up (months)	Complications	Mortality
Darwish et al., 2004 ²⁸	53/F	7	3	Yes	Yes	NR	None	Alive
Ozer et al., 2007 ²⁹	45/F	24	2	Yes	Yes	18	None	Alive
Messerer et al., 2008 ³⁰	69/F	41	3	Yes	Yes	12	None	Alive
Velnar and Bunc, 2008 ³¹	37/F	120	2	Yes	No	NR	NR	NR
Tahir et al., 2008 ³²	48/F	18	2	Yes	No	24	None	Alive
Enwereuzo, et al., 2018 ³³	56/F	98	2	Yes	No	NR	NR	NR
Nawashiro et al., 2019 ²⁴	M/62	NR	2	Yes	GKRS 18 Gy	30	No	Alive
Liu et al., 2020 ²⁰	M/69	72	1	Yes	Yes	9	No	Alive
Abbreviations: GKRS, gamma knife radiosurgery, NR, Not reported.	fe radiosurgery;	NR, Not reported.						

6 | CONCLUSION

Scalp metastasis resulting from atypical meningioma is a rare clinical entity. The common mechanism of metastasis observed in reported cases is the implantation of tumor cells during surgical resection. Therefore, it is imperative for surgeons to adhere to meticulous intra and postoperative surgical practices to minimize the risk of tissue contamination.

AUTHOR CONTRIBUTIONS

Saad Javed: Conceptualization; data curation; writing – original draft. Amina Khan: Data curation; investigation. Ayesha Khalid: Data curation; formal analysis. Gianluca Scalia: Supervision; validation; visualization; writing – review and editing. Giuseppe Emmanuele Umana: Supervision; validation; visualization. Ashraf Mahmood: Methodology; resources; supervision. Eesha Yaqoob: Software; supervision; validation; visualization. Bhavya Pahwa: Supervision; validation; visualization. Bipin Chaurasia: Supervision; validation; visualization.

ACKNOWLEDGMENTS

None.

FUNDING INFORMATION

This manuscript did not receive any funds.

CONFLICT OF INTEREST STATEMENT None.

DATA AVAILABILITY STATEMENT

Data sharing not applicable – no new data generated, or the article describes entirely theoretical research.

ETHICS STATEMENT

This case report was compiled after obtaining informed consent from the patient for the disclosure of clinical history and management with the intention of publication. All attached imaging and clinical materials were deidentified to ensure patient anonymity.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

ORCID

Saad Javed b https://orcid.org/0000-0003-0792-9130 Gianluca Scalia b https://orcid.org/0000-0002-9465-2506 Bhavya Pahwa b https://orcid.org/0000-0002-4010-8951 Bipin Chaurasia b https://orcid.org/0000-0002-8392-2072

Summary of findings of cases included in the review on scalp metastasis due to atypical (WHO grade 2) meningioma.

TABLE 1

REFERENCES

- Ostrom QT, Patil N, Cioffi G, Waite K, Kruchko C, Barnholtz-Sloan JS. CBTRUS statistical report: primary brain and other central nervous system tumors diagnosed in the United States in 2013–2017. *Neuro-Oncology*. 2020;22(12 Suppl 2):iv1-iv96. doi:10.1093/neuonc/noaa200
- Chohan MO, Ryan CT, Singh R, et al. Predictors of treatment response and survival outcomes in meningioma recurrence with atypical or anaplastic histology. *Neurosurgery*. 2018;82(6):824-832. doi:10.1093/neuros/nyx312
- Lüdemann WO, Obler R, Tatagiba M, Samii M. Seeding of malignant meningioma along a surgical trajectory on the scalp. Case report and review of the literature. *J Neurosurg*. 2002;97(3):683-686. doi:10.3171/jns.2002.97.3.0683
- Kok DL, Hendry S, Alvarez B. Iatrogenic subcutaneous metastasis from WHO grade I intracranial meningioma. J Clin Neurosci. 2018;58:224-225. doi:10.1016/j.jocn.2018.09.005
- Lookingbill DP, Spangler N, Sexton FM. Skin involvement as the presenting sign of internal carcinoma. A retrospective study of 7316 cancer patients. *J Am Acad Dermatol*. 1990;22(1):19-26. doi:10.1016/0190-9622(90)70002-y
- Akai T, Shiraga S, Iizuka H, Kishibe M, Kawakami S, Ueda Y. Recurrent meningioma with metastasis to the skin incision case report. *Neurol Med Chir (Tokyo)*. 2004;44(11):600-602. doi:10.2176/nmc.44.600
- Enam SA, Shah MM, Bajwa MH, et al. The Pakistan brain tumour epidemiology study. *J Pak Med Assoc*. 2022;72(Suppl 4, 11):S4-S11. doi:10.47391/JPMA.11-S4-AKUB01
- Champeaux C, Dunn L. World Health Organization grade II meningioma: a 10-year retrospective study for recurrence and prognostic factor assessment. *World Neurosurg*. 2016;89:180-186. doi:10.1016/j.wneu.2016.01.055
- Ostrom QT, Price M, Neff C, et al. CBTRUS statistical report: primary brain and other central nervous system tumors diagnosed in the United States in 2015-2019. *Neuro-Oncology*. 2022;24(Suppl 5):v1-v95. doi:10.1093/neuonc/noac202
- Lagman C, Bhatt NS, Lee SJ, et al. Adjuvant radiosurgery versus serial surveillance following subtotal resection of atypical meningioma: a systematic analysis. *World Neurosurg*. 2017;98:339-346. doi:10.1016/j.wneu.2016.11.021
- Wiemels J, Wrensch M, Claus EB. Epidemiology and etiology of meningioma. J Neuro-Oncol. 2010;99(3):307-314. doi:10.1007/ s11060-010-0386-3
- Shah MM, Khalid MU, Bajwa MH, et al. Meningioma defining characteristics of the affected Pakistani population. J Pak Med Assoc. 2022;72(Suppl 4, 11):S51-S56. doi:10.47391/ JPMA.11-S4-AKUB08
- Avecillas-Chasin JM, Saceda-Gutierrez J, Alonso-Lera P, et al. Scalp metastases of recurrent meningiomas: aggressive behavior or surgical seeding? *World Neurosurg*. 2015;84(1):121-131. doi:10.1016/j.wneu.2015.02.041
- Umana GE, Scalia G, Vats A, et al. Primary extracranial meningiomas of the head and neck. *Life (Basel)*. 2021;11(9):942. doi:10.3390/life11090942
- Ahmed N, Ferini G, Haque M, et al. Primary intraosseous osteolytic meningioma with aggressive clinical behaviour: clinicopathologic correlation and proposed new clinical classification. *Life (Basel)*. 2022;12(4):548. doi:10.3390/life12040548
- 16. Ahmed N, Ferini G, Hossain MATM, et al. Evaluation of surgical cleavage plane by preoperative magnetic resonance

imaging findings in adult intracranial meningiomas. *Life* (*Basel*). 2022;12(4):473. doi:10.3390/life12040473

- Michelson S, Leith JT. Dormancy, regression, and recurrence: towards a unifying theory of tumor growth control. *J Theor Biol*. 1994;169(4):327-338. doi:10.1006/jtbi.1994.1155
- Tohme S, Simmons RL, Tsung A. Surgery for cancer: a trigger for metastases. *Cancer Res.* 2017;77(7):1548-1552. doi:10.1158/0008-5472.CAN-16-1536
- Chiarella P, Bruzzo J, Meiss RP, Ruggiero RA. Concomitant tumor resistance. *Cancer Lett.* 2012;324(2):133-141. doi:10.1016/j.canlet.2012.05.021
- Liu Y, Li J, Duan Y, Ye Y, Xiao L, Mao R. Subcutaneous metastasis of atypical meningioma: case report and literature review. *World Neurosurg*. 2020;138:182-186. doi:10.1016/j.wneu.2020.02.128
- Behling F, Fodi C, Hoffmann E, et al. The role of Simpson grading in meningiomas after integration of the updated WHO classification and adjuvant radiotherapy. *Neurosurg Rev.* 2021;44(4):2329-2336. doi:10.1007/s10143-020-01428-7
- Nanda A, Bir SC, Maiti TK, Konar SK, Missios S, Guthikonda B. Relevance of Simpson grading system and recurrence-free survival after surgery for World Health Organization grade I meningioma. *J Neurosurg.* 2017;126(1):201-211. doi:10.3171/2016.1.JNS151842
- Scheichel F, Popadic B, Kitzwoegerer M, Ungersboeck K, Marhold F. Fluorescence-guided resection in bone and soft tissue infiltrating meningiomas. *Acta Neurochir*. 2020;162(3):605-611. doi:10.1007/s00701-019-04179-7
- Nawashiro H, Nawashiro T, Nawashiro A. Subcutaneous extension of parasagittal atypical meningioma through parietal foramen. *World Neurosurg*. 2019;125:104-105. doi:10.1016/j. wneu.2019.01.185
- Naik V, Pahwa B, Singh M, Kale S, Agrawal D. Correlation based on the WHO grading with tumor control and clinical outcome following gamma knife radiosurgery in meningiomas. *Neurol India*. 2023;71(Supplement):S140-S145. doi:10.4103/002 8-3886.373628
- Song D, Xu D, Han H, et al. Postoperative adjuvant radiotherapy in atypical meningioma patients: a meta-analysis study. *Front Oncol.* 2021;11:787962. doi:10.3389/fonc.2021.787962
- Chun SW, Kim KM, Kim MS, et al. Adjuvant radiotherapy versus observation following gross total resection for atypical meningioma: a systematic review and meta-analysis. *Radiat Oncol.* 2021;16(1):34. doi:10.1186/s13014-021-01759-9
- Darwish B, Munro I, Boet R, Renaut P, Abdelaal AS, MacFarlane MR. Intraventricular meningioma with drop metastases and subgaleal metastatic nodule. *J Clin Neurosci*. 2004;11(7):787-791. doi:10.1016/j.jocn.2004.02.008
- Ozer E, Kalemci O, Acar UD, Canda S. Pin site metastasis of meningioma. *Br J Neurosurg.* 2007;21(5):524-527. doi:10.1080/02688690701485255
- 30. Messerer M, Nouri M, Saikali S, Brassier G, Hamlat A. Métastase sous-cutanée au niveau de la voie d'abord d'un méningiome atypique de la tente du cervelet Cas clinique et revue de la littérature [Subcutaneous metastasis at the operative route of an atypical meningioma of the tentorium. Case report and literature review]. *Neurochirurgie*. 2008;54(4):556-560. doi:10.1016/j. neuchi.2008.02.065
- Velnar T, Bunc G. Iatrogenic metastasis of a benign meningioma to the periosteum at the site of previous craniotomy: a case report. *Wien Klin Wochenschr*. 2008;120(23–24):766-769. doi:10.1007/s00508-008-1068-4

WILEY

- Tahir MZ, Shamim MS, Chishti KN. Recurrent atypical meningioma seeding to surgical scar. *Neurol India*. 2009;57(2):222-224. doi:10.4103/0028-3886.51307
- Enwereuzo C, Moral L, Henneberry JM. Heterotopic cutaneous meningioma: an unusual presentation occurring in a patient with a remote history of intracranial meningioma. *Am J Dermatopathol.* 2018;40(4):e57-e59. doi:10.1097/ DAD.000000000001019

How to cite this article: Javed S, Khan A, Khalid A, et al. Scalp metastasis from atypical meningioma: A case report and literature review. *Clin Case Rep.* 2024;12:e8789. doi:10.1002/ccr3.8789